An Unusual Variant of Congenital Dermal Sinus - A Rare Case Report

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Abstract

Congenital Dermal Sinus tracts (CDS) are most commonly found over the midline and may or may not communicate with the central nervous system. We present a 6-year-old girl with a pit in the right posterior lumbar region noted since birth without any discharge. This location is quite unusual, and our report highlights the variability of congenital dermal sinus tract presentation and raises awareness that, the lesions prone to have a connection with the central nervous system can present quite laterally.

Keywords: Spinal dermal sinus tracts; Spinal dysraphism; Dermal sinus

Introduction

Walker and Busy first coined the term ‘congenital dermal sinus’ in 1934; a rare form of spinal dysraphism occurring during early embryonic life due to incomplete separation of neural ectoderm from epithelial ectoderm. This tract can be located in or near the midline anywhere from the coccyx to the cervical region, with 75% of the tracts observed in the lumbar and lumbo sacral areas. The tract is most often unique, associated with local skin marker.

Case Presentation

A six years old female child was brought to us with the complaint of an abnormal opening in the right posterior lumbar region noted since infancy noticed by the mother while giving bath (Figure 1A). There was no history of any discharge or back pain from the pit. Child was evaluated elsewhere with ultrasonography which revealed sinus tract in the posterior lumbar region without any communication to the spinal canal. Child was referred to our institute for further management.

We evaluated further with Magnetic Resonance Imaging (MRI) which revealed, right posterior lumbar region showing subcutaneous sinus tract measuring 3.5 cm in length and 2 mm maximum thickness in the right posterior abdominal wall, adjacent to iliac crest region without any intra peritoneal or spinal or thecal connection (Figure 2). Underlying muscles were normal. External opening was noted posterolaterally in the subcutaneous plane with tract extending anteromedially and ending in the right posterior paraspinal muscle. Secondary tract was noted anteromedially extending along quadratus lumborum muscle. No intra abdominal or spinal or thecal connection was noted.

Per operatively, we found sinus tract measuring 4 cm × 3 mm, in the subcutaneous plane, ending blindly just above iliac crest (Figure 1B,1C). Surgical excision was done under general anesthesia uneventfully. Histopathology sections showed skin with subcutaneous tissue revealing sinus tract lined by squamous epithelium with the dermis showing a tiny cyst lined by stratified squamous epithelium surrounded by lymphocytes, plasma cells, neutrophils, eosinophils, histiocytes and multinucleate giant cells. Many hair shafts, sebaceous glands and sweat glands were also present in the dermis (Figure 1D,1E). Child is doing well at 8 months follow up.

Discussion

True incidence of congenital dermal sinus is not known; it is estimated to be 1 in every 2500 live births [1,2]. In order of frequency, it is localized most frequently in the lumbo sacral area (41%), followed by the thoracic (10%) and cervical (1%) areas [1,2]. CDS can also be found with other forms of dysraphism with or without any associated cutaneous markers. The CDS tracts can be associated with several pathological findings; including inclusion tumors (for example, epidermoid, dermoid, and teratoma), split-cord malformations and tethered spinal cords [3]. CDS may become symptomatic because of either infection or associated lesions.
Magnetic Resonance Imaging (MRI) is the most specific imaging method to evaluate the spinal canal in neonates and in children, allowing diagnostic confirmation and a more detailed analysis of dysraphism [2-4]. A CDS is lined by a stratified squamous epithelium with lumen containing keratin. There are no published reports of CDS in children, presenting laterally in the lumbar region, without any symptoms, not communicating with spine, in the English literature so far. In view of its rarity, we are reporting this case.

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References